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Children's Yale–Brown Obsessive Compulsive Scale in Autism Spectrum Disorder: Component Structure and Correlates of Symptom Checklist

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Abstract

Objective—Repetitive behaviors in autism spectrum disorders (ASD) range from motor stereotypy to immersion in restricted interests. The modified Children's Yale–Brown Obsessive Compulsive Scale for children with autism spectrum disorder (CYBOCS-ASD) includes a Symptom Checklist (behavior present or absent) and five severity scales (Time Spent, Interference, Distress, Resistance and Control).

Method—We assembled CYBOCS-ASD data from 3 Research Units on Pediatric Psychopharmacology Autism Network trials to explore the component structure of repetitive behaviors in children with ASD. Raters trained to reliability conducted the CYBOCS-ASD in 272 medication-free subjects. Fifteen Checklist items were endorsed for less than 5% of the sample and were dropped. Principal Component Analysis (PCA) was used to explore the clustering of 23 checklist items. Component scores computed for each subject were correlated with other measures. We also examined the distribution of severity scales.

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Results—The subjects (229 boys, 43 girls; mean age = 7.8±2.6 years) met criteria for an ASD; half were intellectually disabled. The PCA resulted in a 5-component solution to classify repetitive behaviors (34.4% of the variance): Hoarding and Ritualistic Behavior; Sensory and Arranging Behavior; Sameness and Self-injurious Behavior; Stereotypy; Restricted Interests. Sensory and Arranging and Stereotypy components were associated with lower adaptive functioning (Pearson r ranged from .2 to .3; $p < 0.003$). The Resistance scale showed little variation with over 60% of sample with the highest score.

Conclusions—Rarely endorsed checklist items can be dropped. The Resistance item does not appear relevant for children with ASD.

Keywords

autism spectrum disorder; clinical trials; outcome measures; repetitive behavior

INTRODUCTION

Autism spectrum disorder (ASD) is a chronic condition of early childhood onset defined by impaired social interaction and social communication as well as repetitive behavior and restricted interests.[1] Current prevalence estimates for ASD range from 0.6 to 1.3% of school-age children in the US.[2,3] Two medications (risperidone and aripiprazole) are currently approved by the US Food and Drug Administration for treating irritability (tantrums, aggression, and self-injury) in children with more narrowly defined autistic disorder. Empirically supported treatments for impaired social interaction or repetitive behavior are less well established and hampered by incomplete consensus on outcome measurement.

Repetitive behavior is a defining feature of other psychiatric disorders including Tourette syndrome (TS) and, obsessive-compulsive disorder (OCD). Tics in TS range from eye blinking, head jerking, and throat clearing to more complex touching habits and blurting out words or parts of words.[4] In OCD, children may show hand-washing, checking or elaborate rituals to prevent harm, or touching in patterns to achieve a sense of completion. [5,6]

Children with ASD may exhibit stereotypic motor behavior such as hand flapping, rocking, spinning objects, or more complex behaviors such as repeatedly watching particular video segments.[7,8] Several investigators have described ‘higher’ and ‘lower’ order repetitive behaviors.[9-12] In this model, higher order behaviors reflect insistence on routines and more purposeful behavior (watching the same video); lower order behaviors include motor stereotypies or sensory-motor behavior such as spinning an object. Other investigators propose circumscribed interests as a third domain.[11] The behavioral correlates of circumscribed interests might include reading books on certain subject or talking about that topic (e.g., mechanical devices) to the exclusion of other topics.[13] In contrast to the unwanted repetitive behaviors in TS and OCD, children with ASD may not struggle against their repetitive behaviors. Indeed, for many children with ASD the repetitive behavior is a preferred activity. For some children, frustration, protest and tantrums may occur when the child is asked to stop the behavior.[7,13]

In preparation for multisite trials in ASD, the Research Units on Pediatric Psychopharmacology (RUPP) Autism Network modified the Children's Yale–Brown Obsessive Compulsive Scale (CYBOCS).[7] The original CYBOCS is a clinician-rated interview designed to measure current symptom severity in youth with OCD.[14] It contains separate checklists for obsessions and compulsions, 5 severity items for obsessions and five for compulsions. The severity items are scored from 0 (not present) to 4 (extreme), yielding an Obsessions score of 0 to 20, a Compulsions score of 0 to 20, and a total of 0 to 40. Given the challenge of ascertaining obsessions in individuals with ASD, the modified CYBOCS for ASD retains only the five compulsion items.[7] We also added items to the compulsions checklist and made minor adjustments to the severity anchor points for use in children with ASD. The revised instrument has demonstrated reliability and validity and has also demonstrated sensitivity to change.[7,15] The purpose of this study is to extend our previous work on adapting the original CYBOCS as an outcome measure in pediatric clinical trials in ASD. To this end, we examined the component structure of the compulsions checklist and the distribution of severity scores on the CYBOCSASD in a sample of well-characterized children with ASD.

METHOD

Participants and Setting

Two hundred ninety one (291) subjects participated in 1 of 3 multisite, randomized trials conducted by the RUPP Autism Network. [16-19] Baseline CYBOCS-ASD checklists were missing or illegible for 19 subjects. Thus, the study sample included 272 subjects (229 boys, 43 girls; aged 4–17 years). The institutional review board at each site approved the studies and written informed consent was obtained from parents or legal guardians prior to data collection. Assent was obtained from children who were able to understand the details of their involvement.

Eligible subjects at baseline were healthy, had a mental age of 18 months or higher, were free of a psychiatric condition requiring another treatment and were medication-free for at least 2 weeks (one month for fluoxetine or antipsychotic medications). In the first trial (RUPP I), 101 subjects with autistic disorder (age 5 to 17 years) accompanied by serious behavioral problems (Aberrant Behavior Checklist [ABC] Irritability subscale ≥ 18) were randomly assigned risperidone or placebo for eight weeks (RUPP Autism Network, 2002). RUPP II compared three doses of methylphenidate to placebo in 66 children (age 5 to 14 years) with autistic disorder, Asperger's disorder or pervasive developmental disorder not otherwise specified (PDD-NOS) plus hyperactivity in a 4-week crossover trial (RUPP Autism Network, 2005). [17] In the third trial (RUPP III), 124 children (age 4.5 to 13 years) with ASD and serious behavioral problems (ABC Irritability subscale ≥ 18) were randomly assigned to risperidone only or risperidone plus parent training for 6 months.[18,19]

Procedures

The clinical assessments were conducted by experienced multidisciplinary teams and included complete medical, developmental, and psychiatric histories, tests of IQ and adaptive functioning, as well as parent interviews and questionnaires.[16-19] The ASD

diagnosis was based on clinical interview, observation and supported by Autism Diagnostic Interview–Revised (Lord *et al.*, 1997).[20]

Measures

Intellectual Functioning—We used one of several standard tests according to the child's ability: Wechsler Intelligence Scale for Children -III; Leiter International Performance Scale–Revised; Mullen Scales of Early Learning; Slosson Intelligence Test; or Stanford-Binet, version 5. Given the use of multiple tests, we classified subjects as intellectually disabled (IQ < 70) or in the normal range (IQ ≥ 70).

Vineland Adaptive Behavior Scales, Survey Edition (Vineland; Sparrow, Balla, and Cicchetti, 1984)—The Vineland is an age and gender standardized measure of adaptive functioning (population mean of 100 ± 15) across three domains: Communication, Socialization, and Daily Living Skills. Vineland items are scored 0 (behavior not performed), 1 (performed sometimes) or 2 (performed on a regular basis).[21]

Aberrant Behavior Checklist (ABC).[22]—This reliable and valid, 58-item informant-based scale (scored 0 to 3) contains 5 factor-analyzed subscales: Irritability, Social Withdrawal, Stereotypy, Hyperactivity, and Inappropriate Speech.

Clinical Global Impression Scale-Severity (CGI-S)[23]—The CGI-S is a commonly used, clinician-rated scale assessing overall impairment. Raters were trained to reliability on this 7-point scale that ranges from non-symptomatic (1) to extreme (7). In these 3 trials; a score of 4 *(Moderate)* or greater was required for entry.

Child and Adolescent Symptom Inventory (CASI).[24]—This 132-item, *DSM-IV*–referenced, parent-rated scale was used to screen for psychiatric disorders. Items are scored from 0 (never) to 3 (very often) and can be scored dimensionally as the sum of the scores for the specific diagnostic subscale (e.g., attention-deficit/hyperactivity disorder [ADHD], depression, etc.). The *CASI-Anxiety scale* is a reliable, 20-item subscale that contains items of generalized anxiety, separation anxiety, social phobia, panic and specific phobia.[265]The *CASI-PDD scale* includes 12 items on social deficits, communication problems, and repetitive behaviors.

Children's Yale–Brown Obsessive Compulsive Scale modified for Autism Spectrum Disorder (CYBOCS-ASD).[7]—The 5-item CYBOCS-ASD is a clinician-rated measure designed to document the current severity of repetitive behavior in children with ASD. The semistructured interview uses the parent as the primary informant, but children are encouraged to participate to the extent possible.[7] The interview includes a Symptom Checklist of possible repetitive behaviors grouped in 8 categories (e.g., Washing Rituals, Checking Behaviors, Hoarding). Raters were trained to mark current symptoms (past week) on the checklist. The absence of a mark indicated that the behavior was *not present*. Each category also permitted the interviewer to write-in “other” behaviors presumed relevant to that overall category. For example, in the section on Repeating Rituals a query about spinning objects might prompt the parent to report “he flips a pencil in front of his eyes.”

Once current repetitive behaviors are identified, the interviewer inquires about Time Spent, Interference in everyday life, Distress (if prevented from performing the behavior), Resistance (child's own effort to limit the behaviors) and degree of Control over the behavior—when instructed by parents to stop the behavior. Each severity item is anchored to the past week and scored from 0 (not present) to 4 (severe) for a total score of 0 to 20.

Rater training included instruction on how to ascertain and document checklist items, identify current target symptoms, and establish current severity. Two to three video recordings were used to illustrate interview methods and scoring. Raters were then asked to score 3 videos independently. Reliable raters had to be within 15% of the gold standard rater on the total score.

Classification of Added Items

All handwritten items added to the checklist for each child were entered verbatim and reviewed by a 2-person panel (A.D., L.S.). Similar items were collapsed to reduce item count and avoid duplication. The resulting item list included 39 items (Table 2). Most items were from the original checklist, some were combinations of original items (e.g., excessive or ritualized hand washing was merged with excessive or ritualized showering, bathing, teeth-brushing, grooming), and others were based on the handwritten items (e.g., watching the same video segment over and over).

Analytic Plan

First, we examined the clinical characteristics of the available sample from the RUPP Autism Network trials and the frequency of symptom endorsement on the CYBOCS checklist. Endorsement was calculated as the number of subjects with that symptom over the total number of subjects.

Principal components analysis (PCA) was used to examine the dimensionality of CYBOCS checklist items. Given that the original CYBOCS checklist was developed for OCD, the PCA proceeded without assumptions about the latent structure of the binary (present or absent) checklist items in children with ASD. To decrease the likelihood of extracting spurious components, we dropped items endorsed by less than 5% of the sample. [26] The number of components extracted was determined to avoid retaining too few (under-extraction) or too many (overextraction) components and to verify sample adequacy.[27,28] Rather than relying on any single criterion (e.g., eigenvalues > 1) for determining the number of components to extract, we considered multiple criteria: the cumulative proportion of item covariance extracted (and the related Scree plot); whether the retained components were well-saturated; and whether the components were well-identified.[28] An orthogonal (varimax) rotation was applied initially and the reliability of the structure was verified using alternate orthogonal (equamax) and oblique (promax) rotations. Rotated item loadings of ≥ 0.3 (moderate effect) were considered salient for interpreting the components, which were subsequently confirmed by content experts (L.S., A.D.). Scores on components were computed for each participant on the basis of all items loadings.

In the third set of analysis, we examined the construct validity of the identified components. These analyses evaluated the association between component scores and scores on

CYBOCS severity scales (Time Spent, Interference, Distress, Resistance, and Control) using Spearman correlation. We also examined the association of component scores with selected subscales on the CASI, ABC subscales and Vineland scores with Pearson correlation. Discriminant function analyses were conducted to determine whether identified components could differentiate participants based on trial-specific treatment targets (hyperactivity in the methylphenidate trial or serious behavioral problems in the risperidone trials), intelligence (IQ ≥ 70 or IQ < 70), or verbal ability (verbal or nonverbal item on the Autism Diagnostic Interview–Revised).

The final set of analysis examined the distributions of the CYBOCS-ASD dimensions (Time Spent, Interference, Distress, Resistance, and Control) and computed the internal consistency (Cronbach's alpha) for all 5 dimensions and with each dimension removed. Pearson correlations were calculated for total CYBOCS-ASD scores with ABC subscales, selected CASI subscales and Vineland score to estimate divergent validity.

RESULTS

Table 1 summarizes the clinical characteristics of the sample. Nineteen subjects were unable to cooperate with testing and did not have an IQ score. Of those with IQ data, 141 (51.8%) were classified as intellectually disabled (IQ < 70). Based on parent-report, the sample was 72.4% Caucasian, 12.5% African-American, 7.4% Hispanic, 5.1% Asian, and 2.6% identified as ‘other.’

Fifteen checklist items were endorsed for less than 5% of subjects and were dropped. As shown in Table 2, the rate of endorsement for the remaining 24 items ranged in frequency from 7% (ordering, putting objects into proper place) to 62% (body stereotypy).

Principal Components Analysis

Twenty-four items were initially included in the PCA, however 1 item (excessive ritualized hand washing, showering) did not reach our threshold (≥.30) on any component and was dropped. Twenty-three checklist items were included in a second PCA (16 original items and 7 new items). The median endorsement rate for these 23 items was 18%. This frequency is 11 points above the minimum and 20% of the 55-point range (7 to 62%) suggesting that pairwise disparities in marginal frequencies were not extreme and mitigates concerns regarding binary item data. A 5-component solution provided the best classification of repetitive behaviors (accounting for 34.4% of the variance). Four and six component solutions were considered. The four component solution accounted for less variance; the 6-component solution resulted in a higher number items loading on more than 1 component.

Table 3 displays the varimax rotated component structure, which was consistent across the alternative equamax and promax rotations. The 5-component model yielded a different structure than the original CYBOCS categories. Component I included Hoarding and Ritualistic Behavior (e.g., counting, ritualized eating, and checking); Component II involved Sensory/Motor (e.g., echolalia, repetitive water play) and Arranging Behaviors (e.g., ordering or lining up objects); Component III included Insistence on Sameness and Self-Injurious Behaviors (SIB); Component IV comprised Stereotypy; and Component V

captured Restricted Interests. Object Stereotypy (e.g., repetitive manipulation of a pencil, straw, piece of string) loaded on Components III (Sameness) and IV (Stereotypy). Given the minimal difference on loadings of object stereotypy for Components III and IV, we placed this item on both components. A few other items showed similar loadings on more than 1 Component (Repetitive Sexual Behavior loaded on Component II (Sensory and Arranging) and Component III (Sameness and SIB). Hair-pulling loaded on Components I and V. Components I and III, blinking and staring rituals also loaded on Components I and IV. By contrast, there was a higher loading for hoarding on Component I compared to Component V (Restricted Interests), suggesting that hoarding belongs in Component I. Perhaps fascination with a specific object (Component V) drives the reluctance to discard the object —rather than hoarding as a general problem.

Association of the 5 Components to Other Measures

Based on parent report, many subjects exhibited repetitive behavior in more than one component. Standardized scores (mean = 0, SD = 1) on each of the 5 components were computed for each participant. A higher component score for a given subject reflects greater likelihood of exhibiting that type of repetitive behavior. Table 4 presents correlations between component scores and age as well as scores on several clinical measures.

Subjects ranked with higher scores on the CYBOCS-ASD Distress dimension were higher on Component I (Ritualized behavior) (Spearman rho = .31, $p < 0.003$) (see Table 4). Scores on the CASI Anxiety scale were associated with scores on Component I (Pearson r = .30, $p < 0.003$). Subjects with higher scores on Component II scored higher on the CASI-PDD scale (r = .33, $p < 0.003$). Although significant, these small to medium correlations suggest that components are not strongly associated with clinical measures.

Discriminant function analyses demonstrated that the five component scores did not discriminate on trial-specific targets (hyperactivity in the methylphenidate trial or serious behavioral problems in risperidone trials; Lambda=0.964, p=0.081). However, component scores differentiated significantly by IQ (Lambda=0.826, p<0.001). Participants with lower IQ had lower scores on Component 1 (Hoarding and Ritualistic Behavior) (p=0.005) and higher scores on Component II (Sensory and Rule Based Behavior) and Component IV (Stereotypy) (p<0.001 for both). Similarly, these three component scores differentiated among nonverbal versus verbal subjects (based on the Autism Diagnostic Interview–Revised [ADI-R]) (Lambda=0.854, p<0.001). Participants who were nonverbal had lower scores on Component I (p=0.005) and higher scores on Component II (p=0.021) and Component IV (p<0.001). Component III (Sameness and SIB) and Component V (Restricted Interest) were not significant in any discriminant analyses.

Distribution of Scores on CYBOCS-ASD Dimensions

The Resistance and Control dimensions each had a modal score of 4 compared to a modal score of 3 for the other 3 dimensions (Table 5). The distribution of the Resistance item is particularly distorted with over 60% of subjects rated extreme (score of 4).

The internal consistency for the 5-item CYBOCS-ASD Total score was .81. When each item was removed, Cronbach's alpha ranged from .76 to .79 (see Table S1, available online). The CYBOCS-ASD total score showed modest correlations with measures of adaptive functioning and maladaptive behavior. For example, correlations with the ABC subscales for Irritability, Social Withdrawal, Stereotypy, and Hyperactivity ranged from .25 to .32 ($p < .01$). Similarly, correlations between the CYBOCS-ASD total score and the Vineland (higher scores reflect higher functioning) were also modest, (−.26 to −.30, $p < .01$ for all three domains) (see Table S2, available online). These small to medium correlations suggest that the CYBOCS-ASD is measuring a separate construct from these other instruments.

DISCUSSION

In its current form, the CYBOCS-ASD is a reliable and valid measure of repetitive behavior in youth with ASD.[7] The results of the current study show that several original CYBOCS checklist items are not relevant for children with ASD and can be dropped. Twenty-three checklist items endorsed by at least 5% of this sample fell into five components. These five clinically plausible components explained nearly 35% of the variance. For example, Component IV (Stereotypy) includes motor behaviors such as repetitive object manipulation, hand and finger stereotypy, blinking and staring rituals, and repetition of routine activities. In this sample, subjects often exhibited behaviors in more than one component suggesting that typologies of repetitive behavior in ASD may not be strictly drawn. By contrast, however, the discriminant function analysis showed that children with verbal capacity (as reported on the ADI-R) and those with IQ ≥ 70 were more likely to exhibit counting, checking, and ritualized eating behaviors (Component I) than nonverbal children or those with intellectual disability. Lower functioning children were more likely to engage in ordering and arranging, hand and finger stereotypy, and object manipulation (Components II and IV). Collectively, these findings support the format of the CYBOCS-ASD, which yields a total score based on the impact of the symptoms present rather than subscale scores for types of repetitive behavior. A reliable and valid clinician-rated measure with a single total score has obvious statistical advantages as an endpoint in clinical trials.

The components identified in this analysis show differences and similarities to previous investigations. The International Classification of Disease–Tenth edition (ICD-10) describes 4 categories of repetitive behavior in ASD: (a) preoccupations with part-objects; (b) stereotyped motor mannerisms; (c) circumscribed patterns of interest; (d) adherence to specific routines or rituals.[29] Several prior factor analyses based on the 10–12 repetitive behavior items from the ADI-R reported 2 factors: stereotypy and insistence on sameness.[30] This is consistent with the traditional concepts of ‘lower’ and ‘higher’ order behaviors.[9] However, circumscribed interests were not included in most of these ADI-R factor analysis. One study that did include circumscribed interest showed that it emerged as a third factor, which is consistent with ICD-10.[11] The limited number of repetitive behaviors on the ADI-R may have hindered the identification more than 2 factors.

Honey *et al.*[12] examined the factor structure of the 26-item, parent-rated Repetitive Behavior Questionnaire (RBQ)[9] in 180 children with ASD age 3 to 16 years. Children with self-injurious behavior (SIB) were excluded from the analysis. Two factors emerged: Sensory/

Motor behaviors and Insistence on Sameness/ Circumscribed Interests. This classification merges the four ICD-10 categories into two. Preoccupations with part-objects and stereotyped motor mannerisms formed a single Sensory/Motor factor; circumscribed patterns of interest and adherence to routines became a single Sameness/Circumscribed Interest factor. This 2-factor solution may have missed the distinction between sameness and circumscribed interests reported by Lam *et al.* and the findings in the present study.[11]

Five factors have been identified for the parent-rated Repetitive Behavior Scale–Revised (RBSR) developed by Bodfish *et al*.[10,31,32] These factors do not precisely correspond with ICD-10. For example, it includes an SIB factor not listed in ICD-10. As in ICD-10, the RBSR has separate factors for stereotypic behavior, insistence on sameness and circumscribed interests. The fifth factor included ordering and arranging and the need to touch or tap things is similar to our Component II—but not clearly enumerated in ICD-10.

The 5-component structure identified in the current study included a larger set of behaviors than the ADI-R. Unlike the study of the RBQ by Honey *et al.*,[12] our study included SIB. Component III in our analysis includes insistence on routines and SIB—which does not fit neatly with the concept of higher and lower order repetitive behaviors,[9] but has been reported by others.[35] Although SIB is multidetermined, it may be that some children resort to SIB when routines are not followed. Consistent with the concept of lower order repetitive behaviors, Components II (Sensory Motor) and IV (Stereotypy) in this study were associated with nonverbal and intellectually disabled subjects. However, these 2 components emerged as distinct in our analysis—rather than the single factor reported by others.

Taken together, repetitive behaviors exhibited by children with ASD appear to cluster in patterns that are collapsed in some analyses and more finely delineated in others. Other seeming discrepancies in factor analysis may be due to differences in sample characteristics, sample size, as well as differences in the coverage of behaviors. Children with ASD often exhibit behaviors from more than one category suggesting that repetitive behaviors are not factor-specific. The association of motor stereotypy and sensory-motor behavior with intellectual disability and the association of more complex ritualistic behavior with IQ ≥ 70 are replicated findings.[10,11] The implications for treatment are not clear.

Consistent with the results of our previous report, this study raises questions about the relevance of the Resistance item in the ASD population.[7] In OCD, resistance may be a sign of health. Patients who resist the urge to carry out ritualized behavior are considered less severe on this dimension than those who do not resist.[34] Given that individuals with ASD often engage in repetitive behavior as a preferred activity, resistance is less informative. Indeed, over 60% of the subjects in our sample had a Resistance score of 4, reflecting no resistance. Whether the Resistance item score changes with treatment is an unanswered empirical question. If the Resistance item is dropped, additional severity items such as intensity of the behavior or the unusual appearance of the behavior could be developed and tested to retain the scoring range from 0 to 20.

Our results underscore other differences in repetitive behavior in ASD compared to OCD. Repetitive behaviors in OCD such as excessive hand washing and rituals to prevent harm

observed in children with OCD were rarely endorsed in this sample of children with ASD. In practice, attention to these distinctions may help with differential diagnosis. The phenomenological dissimilarities in OCD and ASD may explain the differential response to the selective serotonin reuptake inhibitors .[35]

The results of this study provide an incremental refinement on measuring repetitive behavior in children with ASD. However, there are several limitations of this study. First, this was a sample of convenience consisting of children selected for participation in multisite clinical trials. The sample may not be representative of all children with ASD. Second, the symptom checklist included a wide range of repetitive behaviors generated by an expert panel and compulsions from the original CYBOCS. Nonetheless, we may have missed some repetitive behaviors in children with ASD. Future research could explore whether the addition of relevant repetitive behaviors affects the component structure. Third, we used the simple exploratory model (PCA) in this study without assumptions about the latent structure of the binary (present or absent) checklist items. Future studies could proceed with confirmatory analysis to verify this component structure in youth with ASD. Fourth, the weights used to generate component scores, which were then correlated with other measures are sample-specific. These associations may change in magnitude in a different sample.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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References

1. American Psychiatric Association. , editor. DSM5: Diagnostic and Statistical Manual of Mental Disorders. Fifth Edition.. American Psychiatric Association; Washington, DC: 2013.
2. Centers for Disease Control and Prevention. Prevalence of Autism Spectrum Disorders: Autism and Developmental Disabilities Monitoring Network, 14 Sites, United States, 2008. Morbidity and Mortality Weekly Report. Surveillance Summaries. 2012; 61(3)
3. Fombonne E. The epidemiology of pervasive developmental disorders. Pediatric Research. 2009; 65(6):591–8. [PubMed: 19218885]
4. Leckman JF, Bloch MH, Scahill L, King RA. Tourette syndrome: the self under siege. J Child Neurology. 2006; 21(8):642–9.
5. Mataix-Cols D, Nakatani E, Micali N, Heyman I. Structure of obsessive-compulsive symptoms in pediatric OCD. J Amer Acad Child Adolesc Psychiatry. 2008; 47(7):773–8. [PubMed: 18344900]
6. Rosario-Campos MC, Miguel EC, Quatrano S, Chacon P, Ferrao Y, Findley D, et al. The Dimensional Yale-Brown Obsessive-Compulsive Scale (DY-BOCS): an instrument for assessing obsessive-compulsive symptom dimensions. Molecular Psychiatry. 2006; 11(5):495–504. [PubMed: 16432526]
7. Scahill L, McDougle CJ, Williams SK, Dimitropoulos A, Aman MG, McCracken JT, et al. The Children's Yale-Brown Obsessive Compulsive Scales modified for pervasive developmental disorders. J Amer Acad Child Adolesc Psychiatry. 2006; 45(9):1114–23. [PubMed: 16926619]
8. Zandt F, Prior M, Kyrios M. Repetitive behavior in children with high functioning autism and obsessive-compulsive disorder. J Autism Develop Disorders. 2007; 2007; 37:251–259.
9. Turner M. Annotation: Repetitive Behaviour in Autism: A Review of Psychological Research. J Child Psych Psychiatry. 1999; 40(6):839–849.
10. Bodfish JW, Symons FJ, Parker D, Lewis MH. Varieties of repetitive behavior in autism:comparisons to mental retardation. J Autism Developl Disorders. 2000; 2000; 30:237–243.
11. Lam KSL, Bodfish JW, Piven J. Evidence for three subtypes of repetitive behavior in autism that differ in familiality and association with other symptoms. J Child Psych Psychiatry. 2008; 2008; 49(11):1193–1200.
12. Honey E, McConachie H, Turner M, Rodgers J. Validation of the repetitive behaviour questionnaire for use with children with autism spectrum disorder. Res Autism Spectrum Disorders. 2012; 6:355–364.
13. Turner-Brown LM, Lam KSL, Holtzclaw TN, Dichter GS, Bodfish JW. Phenomenology and measurement of circumscribed interests in autism spectrum disorders. Autism. 2011; 2011; 15(4): 437–456. [PubMed: 21454386]
14. Scahill L, Riddle MA, McSwiggin-Hardin M, Ort SI, King RA, Goodman WK, et al. Children's Yale-Brown Obsessive Compulsive Scale: reliability and validity. J Amer Acad Child Adolesc Psychiatry. 1997; 36(6):844–852. [PubMed: 9183141]
15. McDougle CJ, Scahill L, Aman MG, McCracken JT, Tierney E, Davies M, et al. Risperidone for the Core Symptom Domains of Autism: Results from the RUPP Autism Network Study. Amer J Psychiatry. 2005; 162:1142–1148. [PubMed: 15930063]
16. Research Units on Pediatric Psychopharmacology Autism Network. Risperidone in children with autism for serious behavioral problems. New Eng J Medicine. 2002; 347(5):314–321.
17. Research Units on Pediatric Psychopharmacology (RUPP) Autism Network. Randomized, controlled, crossover trial of methylphenidate in pervasive developmental disorder. Archives of General Psychiatry. 2005; 62:1266–74. [PubMed: 16275814]
18. Aman MG, McDougle CJ, Scahill L, Handen B, Johnson C, Stigler KA, et al. Medication and parent training in children with pervasive developmental disorders and serious behavioral problems: Results from a randomized clinical trial. J Am Acad Child Adolesc Psychiatry. 2009; 2009; 48(12):1143–54. [PubMed: 19858761]
19. Scahill L, McDougle CJ, Aman MG, Johnson C, Handen B, Bearss K, et al. Effects of risperidone and parent training on adaptive functioning in children with a pervasive developmental disorders and serious behavioral problems. J Am Acad Child Adolesc Psychiatry. 2012; 51(2):136–146. [PubMed: 22265360]

20. Lord C, Pickles A, McLennan J, Rutter M, Bregman J, Folstein S, Fombonne A, Leboyer M, Minshew N. Diagnosing autism: analyses of data from the Autism Diagnostic Interview. Journal of Autism and Developmental Disorders. 1997; 27:501–517. [PubMed: 9403369]
21. Sparrow, SS.; Balla, DA.; Cicchetti, DV. Vineland Adaptive Behavior scales: Survey form manual. American Guidance Service; Circle Pines: 1984.
22. Aman MG, Singh NN, Stewart AW, Field CJ. Psychometric characteristics of the Aberrant Behavior Checklist. Am J Ment Defic. 1985; 89:492–502. [PubMed: 3158201]
23. Guy, W. ECDEU assessment manual for psychopharmacology. DHEW, NIMH; Washington, DC: 1976.
24. Gadow, KD.; Sprafkin, J. Child symptom inventory-4 screening and norms manual. Checkmate Plus; Stony Brook, NY: 2002.
25. Hallett V, Lecavalier L, Sukhodolsky DG, Cipriano N, Aman MG, McCracken JT, et al. Exploring the manifestations of anxiety in children with Autism Spectrum Disorders. J Autism Dev Disord. 2013; 43(10):2341–52. [PubMed: 23400347]
26. Gorsuch, RL. Factor Analysis. Erlbaum; Hillsdale, NJ: 1983. 1983.
27. Wood JM, Tataryn DJ, Gorsuch RL. Effects of under- and overextraction on principal axis factor analysis with varimax rotation. Psychological Methods. 1996; 1:354–365.
28. Velicer WF, Fava JL. Effects of variable and subject sampling on factor pattern recovery. Psychological Methods. 1998; 3:231–251.
29. World Health Organization. The ICD-10 classification of mental and behavioural disorders: Diagnostic criteria. WHO; Geneva: 2007.
30. Honey E, Rodgers J, McConachie H. Measurement of restricted and repetitive behaviour in children with autism spectrum disorder: Selecting a questionnaire or interview. Res Autism Spectrum Disorders. 2012; 6:757–776.
31. Lam KSL, Aman MG. The Repetitive Behavior Scale–Revised: Independent Validation in Individuals with Autism Spectrum Disorders. Journal of Autism and Developmental Disorders. 2007; 37(5):855–866. [PubMed: 17048092]
32. Bishop SL, Hus V, Duncan A, Huerta M, Gotham K, Pickles A, Kreiger A, Buja A, Lund S, Lord C. Subcategories of restricted and repetitive behaviors in children with autism spectrum disorders. Journal of Autism and Developmental Disorders. 2013; 43(6):1287–1297. [PubMed: 23065116]
33. Duerden EG, Oatley HK, Mak-Fan MK, McGrath PA, Taylor MJ, Szatmari P, Roberts SW. Risk factors associated with self-injurious behaviors in children and adolescents with autism spectrum disorders. Journal of Autism and Developmental Disorders. 2012; 42:2460–2470. [PubMed: 22422338]
34. GoodmanWK, Price LH.; Rasmussen, SA.; Mazure, C.; Fleischmann, RL.; Hill, CL., et al. Yale-Brown Obsessive Compulsive Scale. I. Development, use, and reliability. Arch Gen Psychiatry. 1989; 46(11):1006–11. [PubMed: 2684084]
35. King BH, Hollander E, Sikich L, McCracken JT, Scahill L, Bregman JD, et al. For the STAART Psychopharmacology Network. Lack of efficacy of citalopram in children with autism spectrum disorders and high levels of repetitive behavior. Arch Gen Psychiatry. 2009; 66(6):583–590. [PubMed: 19487623]

Table 1

Clinical Characteristics of 272 subject in 3 Research Units on Pediatric Psychopharmacology (RUPP) Autism Network Trials

Subject variable	
Gender, male, n (%)	229 (84.2)
< IQ 70[a], n (%)	141(55.7)
Functional Language (on ADI-R verbal item), n (%)	198 (73.6)
Diagnosis, n (%)	
Autistic Disorder	209 (76.8)
Asperger's Disorder	14 (5.1)
PDD-NOS	49 (18.0)
Clinical Global Impression Severity, n (%)	
Moderate	75 (27.7)
Marked	135(49.8)
Severe	59 (21.8)
Extreme	2 (.7)
Age (years), m (SD)	7.8 (2.6)
CYBOCS-ASD Total, m (SD)	14.8 (3.2)
Time spent	2.9 (.94)
Interference	2.4 (.91)
Distress	2.7 (.86)
Resistance	3.4 (.79)
Control	3.3 (.75)
Child and Adolescent Symptom Inventory (CASI), m (SD)	
Anxiety Scale	14.7 (9.5)
PDD scale	22.2 (7.6)
Vineland Adaptive Behavior Scales, m (SD)	
Communication	54.9 (20.9)
Socialization	55.6 (15.9)
Daily Living	46.1(20.4)
Aberrant Behavior Checklist, m (SD)	
Irritability	25.4 (9.3)
Social Withdrawal	15.0 (8.9)
Stereotypy	8.7 (5.4)
Hyperactivity	34.1 (8.7)
Inappropriate Speech	5.9 (3.8)

Note: ADI-R=Autism Diagnostic Interview–Revised; CASI= Child and Adolescent Symptom Inventory; CYBOCS-ASD=Children's Yale–Brown Obsessive Compulsive Scale for children with autism spectrum disorder; PDD-NOS=pervasive developmental disorder not otherwise specified.

[a] IQ was missing for 19 subjects due to poor cooperation with testing

Table 2

Children's Yale–Brown Obsessive Compulsive Scale for Children With Autism Spectrum Disorder (CYBOCS-ASD) Symptom Checklist Items Prior to Principal Component Analyses

Checklist Category	Checklist Items	Endorsement, n (%)
Washing/Cleaning:	Excessive or ritualized hand washing, showering, bathing[a]	22 (8.1)
	Excessive cleaning of items (e.g., personal clothing)	**13 (4.8)**
	Repetitive water play	92 (33.8)
	Other measures to prevent/remove contaminants	**7 (2.6)**
Checking:	Checking locks, toys, items, etc.	49 (18)
	Checking associated with getting washed, dressed, or undressed	**11 (4)**
	Checking that did not/will not harm others	**1 (.4)**
	Checking that did not/will not harm self	**0 (0)**
	Checking that nothing terrible did/will happen	**7 (2.6)**
	Checking that did not make mistake	**6 (2.2)**
	Checking tied to somatic obsessions	**3 (1.1)**
Repeating:	Rereading, (e.g., telephone book, train schedule) or rewriting	35 (12.9)
	Need to repeat routine activities	38 (14)
	Touching in patterns	27 (9.9)
	Body stereotypy[a]	168 (61.8)
	Object stereotypy[a]	75 (27.6)
	Echolalia	133 (48.9)
	Hand, finger stereotypy[a]	40 (14.7)
	Repetitive behavior involving sensation[a]	43 (15.8)
	Restricted interests (i.e.,extended play with preferred object, watching same video over and over)[a]	79 (29)
Counting:	Counting objects, certain numbers, words, etc.	22 (8.1)
Ordering/Arranging:	Lining up objects or evening up	132 (48.5)
	Ordering, arranging (insistent on placement of toys/objects/furniture in assigned place)[a]	19 (7)
Hoarding/Saving:	Difficulty throwing things away, saving bits of paper, string	66 (24.3)

Checklist Category	Checklist Items	Endorsement, n (%)
Excessive Games/Superstitious	**Excessive games/superstitious behavior**	**9 (3.3)**
Behavior:		
Rituals involving another person:	Need to involve another person(usually a parent) in ritual	129 (47.4)
	Repetitive requests or demands	**2 (.7)**
	Dwelling on a preferred topic (not echolalia)[a]	**12 (4.4)**
	Insistent on routines in everyday situations[a]	56 (20.6)
Miscellaneous:	**Mental rituals**	**2 (.7)**
	Need to tell, ask, or confess	29 (10.7)
	Measures to prevent harm to self; or terrible consequences	**0 (0)**
	Ritualized eating behaviors	66 (24.3)
	Repetitive sexual behavior	41 (15.1)
	Excessive list making	**2 (.7)**
	Rituals involving blinking or staring	32 (11.8)
	Trichotillomania, skin picking	48 (17.6)
	Self-injurious behavior	54 (19.9)
	Pica [a]	**1 (.4)**

Note: Bold indicates items endorsed by less than 5% of subjects dropped for the Principal Component Analysis (PCA).

[a]Compiled items are combinations of the original items or newly created items based on the “other” items reported.

Table 3

Varimax Rotated Component Structure of Repetitive Behaviors on the Children's Yale–Brown Obsessive Compulsive Scale for Children With Autism Spectrum Disorder Symptom Checklist (N = 272)

Checklist Items	Frequency	I Hoarding and Ritualistic Behavior	II Sensory and Arranging	III Sameness/SIB	IV Stereotypy	V Restricted Interest
Difficulty throwing things away, saving bits of paper, string	24.3	**.536**	–.057	–.054	–.204	**.322**
Counts objects, certain numbers, words, etc.	8.1	**.527**	.027	.022	.048	–.076
Checks locks, toys, books/items, etc.	18.0	**.520**	–.173	.247	.129	.081
Ritualized eating behaviors	24.3	**.513**	.047	–.025	–.033	–.209
Trichotillomania (hair-pulling), skin picking	17.6	**.389**	.111	.110	.034	**.321**
Ordering, arranging (placing toys/objects/furniture in assigned place)[a]	7.0	–.213	**.639**	.210	–.043	.043
Echolalia	48.9	.293	**.581**	–.076	–.101	.001
Repetitive behavior involving sensation[a]	15.8	–.048	**.473**	–.094	–.118	.261
Repetitive water play	33.8	.088	**.454**	–.027	.262	–.224
Repetitive sexual behavior	15.1	–.076	**.350**	**.373**	.231	.156
Rereading or rewriting	12.9	.119	–.066	**.607**	–.133	–.234
Insisting on routines[a]	20.6	–.011	.004	**.563**	–.104	.214
Self-injurious behavior	19.9	.206	.280	**.442**	.111	–.011
Body stereotypy[a]	61.8	–.078	.247	**–.371**	.125	–.101
Object stereotypy[a]	27.6	–.140	.102	**.340**	**.383**	.040
Need to repeat routine activities	14.0	.213	.087	–.031	**.483**	.123
Hand/finger stereotypy[a]	14.7	.053	–.083	–.183	**.349**	.054
Rituals involving blinking or staring	11.8	**.307**	–.032	.092	**.366**	.193
Need to involve another person in ritual	47.4	**.379**	.183	.009	**–.591**	.093
Need to tell, ask, or confess	10.7	.052	–.060	.046	**–.463**	.188
Touching in patterns	9.9	.073	–.031	.077	–.031	**.597**
Restricted interests (i.e., trains, videos, etc.)[a]	29.0	.008	.157	.094	.082	**.348**
Need for symmetry or evening up	48.5	**.332**	.122	**.310**	.033	**–.538**

Note: Loadings with an absolute value ≥ .30 are in bold. Underlined items load similarly on 2 Components. The 5 Components accounted for 34.4% of the variance.

[a]Compiled items.

Table 4

Correlations Between Principal Component Scores and Scores on Clinical Measures

	I Hoarding/Ritualistic Behavior	II Sensory and Arranging	III Sameness/SIB	IV Stereotypy	V Preoccupation
Age	.03	.05	–.02	–.12	.23
CYBOCS-ASD Total Severity[a]	**.28***	**.27***	.04	.09	–.07
Time spent	**.23***	**.25***	–.02	.13	–.01
Interference	.18	**.26***	.02	.10	–.05
Distress	**.31***	.16	.02	.06	–.15
Resistance	.10	.11	.05	–.05	.02
Control	.18	.10	.06	.03	.02
CASI					
Anxiety Scale	**.30***	.06	–.05	–.06	.02
PDD scale	.03	**.33***	.01	.18	–.01
Vineland Adaptive Behavior Scale[b]					
Communication	.09	**–.26***	.03	**–.31***	–.06
Socialization	–.01	**–.21***	–.07	–.15	–.11
Daily Living	.02	**–.21***	–.01	–.17	–.10
Aberrant Behavior Checklist					
Irritability	**.21***	.14	.07	**.20***	–.02
Social Withdrawal	.08	**.21***	.01	.14	–.04
Stereotypy	–.05	**.28***	–.16	**.24***	–.01
Hyperactivity	.07	.07	–.06	.04	–.07
Inappropriate Speech	.17	.13	–.04	–.13	–.02

Note: CASI= Child and Adolescent Symptom Inventory; CYBOCS-ASD= Children's Yale–Brown Obsessive Compulsive Scale for children with autism spectrum disorder; PDD = pervasive developmental disorder; SIB = self-injurious behavior.

[a] Spearman, all others Pearson

* $p<.003$

[b] Higher Vineland scores reflect better adaptive functioning, hence the negative correlations

Table 5

Item Mean, SD, and Frequency Distribution of Children's Yale–Brown Obsessive Compulsive Scale for Children With Autism Spectrum Disorder (CYBOCS-ASD) Severity Dimensions

	Time spent	Interference	Distress	Resistance	Control
m (SD)	2.86 (.94)	2.43 (.91)	2.73 (.86)	3.45 (.79)	3.32 (.75)
Frequency Distribution, n (%)					
0 (none)	2 (.7)	7 (2.6)	6 (2.2)	2 (.7)	2 (.7)
1 (mild)	24 (8.8)	33 (12.1)	18 (6.6)	3 (1.1)	3 (1.1)
2 (moderate)	58 (21.3)	92 (33.8)	57 (21.0)	30 (11.0)	26 (9.6)
3 (severe)	115 (42.3)	115 (42.3)	154 (56.6)	73 (26.8)	116 (42.6)
4 (extreme)	73 (26.8)	25 (9.2)	37 (13.6)	164 (60.3)	125 (46.0)